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WHAT HAS BEEN THE EFFECT ON TRIAL OUTCOME ASSESSMENTS OF A DECADE OF PATIENT PARTICIPATION IN OMERACT?

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ABSTRACT

Objective Since 2002 58 patients have participated as collaborating partners in 6 Outcome Measures in Rheumatology (OMERACT) conferences. Little is known about how they engage with researchers and how they have influenced the conference outcomes.

Method A responsive evaluation was carried out, including a thematic document analysis of conference proceedings and grey literature, participant observation, and 38 interviews with patients and professionals representing research, industry and regulators. Interview transcripts were subjected to an inductive content analysis.

Results The role of patients has evolved from a single focus group in 2002 to full integration in all parts of the conference in 2012. Long term engagement has made a significant change in the scope and conduct of rheumatology research. It has enriched the research agenda by identifying previously neglected outcome domains such as fatigue, sleep disturbances and flares, and it has contributed to more patient relevant outcomes in clinical trials. Facilitating factors have been the strong commitment of the leadership; an adequate selection procedure; an inclusive conference design; an interactive and encouraging moderation style, and self organized support. The intensity of the program and doubts regarding the representativeness of the patient group were still seen as challenges for the future.

Conclusions Making patient participation an integral part of the vision and procedures of OMERACT has significantly contributed to the success of OMERACT. It has changed the perceptions and beliefs of many participants. Full utilization of patients' experiential knowledge before and during the conference is still challenging.

Introduction

There is a growing interest in actively engaging with patients in the context of scientific research.[1 2] Involving patients in the development of patient reported outcomes (PROs) ensures their relevance, acceptability and quality.[2-4] Outcome Measures in Rheumatology (OMERACT) has been a pioneer the field of rheumatology in organizing long-term involvement of patients as collaborating partners[5-8] in its bi-annual, worldwide conferences. Since 2002, 58 patients with different rheumatic conditions have participated in six conferences with the purpose of providing the patient perspective.[9] Their involvement has been reported as advantageous, a 'success'[10] and 'the beginning of a paradigm shift.'[11] In 2011 OMERACT formalized its policy towards patient participation, recognizing the essential role of patients in outcome research.[12] In this, patients' input is indispensible when identifying domains that are important from the perspective of patients and assessing feasibility of measurement tools, and provides face validity to the OMERACT process. It also enables OMERACT to ground theoretical discussions "in the lived experience of arthritis, and in concepts which can be readily communicated to patients to help with therapeutic decision making."[12]

Despite a widespread belief among experienced OMERACT delegates that patient participation is worthwhile, robust evidence for its effectiveness is lacking. Little is known in general about how patients engage with researchers, the specific features which promote collaborative research, and the overall impact and consequences of patient involvement. OMERACT, because of its long history of patient participation, provides a unique opportunity to explore these issues by analyzing the impact of patient participation on the nature of its research activities. In addition, the inhibiting and facilitating conditions for this process can be elucidated.

This article contains the preliminary findings of a PhD project as presented by the first author during OMERACT 11 (2012). We describe the combined results of a thematic document analysis of conference proceedings and 38 interviews during OMERACT 10 (May 2010, Malaysia) based on three empirical sub-studies.[13-15]

OMERACT 10 took place over six days at a residential conference center and focused on four diseases – rheumatoid arthritis (RA), psoriatic arthritis, ankylosing spondylitis and gout - and on the methodology of choosing domains of interest and instruments appropriate to measuring these domains. As in previous meetings, the program consisted of a mixture of plenary sessions, intense small group discussions and, towards the end, formal presentations and proposals which were voted on to achieve international consensus.[16] Twenty of the 200 places were allocated to patients.

Methods

To assess the process and impact of patient involvement in OMERACT, a responsive evaluation [17 18] was carried out, starting with a thematic document analysis including scientific literature on patient participation, OMERACT conference proceedings and grey literature such as correspondence, invitations, session reports and policy documents. The review focused on the reception and evolution of patient involvement in OMERACT conferences and the contributions made by patients.

Data collection was based on 38 semi-structured interviews before, during and after OMERACT 10 (Malaysia, 2010), and included researchers (n=12), representatives of pharmacological industry, regulators and staff (n=4), experienced patients (n=8) and new patients (n=8). To gain insights into changing perceptions, three new patients were interviewed three times: before the start of the conference; on the third day; and immediately after the end. Finally the opinions and experiences of 16 professionals and 16 patients were collected (See table 1 in chapter 5).

Selection of interviewees aimed at maximum variation and followed an emergent purposive sampling approach taking into account stakeholder background, opinion about patient involvement, gender, geographical spread and number of OMERACT conferences attended. A preliminary analysis of the first data revealed an unexpected response shift. The attitude towards patient involvement of two professionals had changed from presumed skeptic to supportive. For this reason we identified two new interviewees who were known for their critical perceptions. After analyzing the additional transcripts, saturation was achieved.

Broad interview protocols were developed[19] based on the document analysis, four pilot interviews, and the first author's personal experience during three OMERACT conferences. The protocols differed for professionals, new patients and experienced patients (Table 1). The interviews were recorded, transcribed and subjected to a responder check. One interview took place without protocol and, on request of the interviewee, without recording. The average duration of the interviews was about 50 minutes. Finally, participant observation took place during six patient meetings by the first author.

Interview transcripts were subjected to an inductive content analysis.[20] Coding of all interviews was done separately by the first author and an independent health researcher with extensive experience in qualitative research (MK). To increase the relevance and validity of the data analysis one of the patients who attended OMERACT 10 for the first

Table 1 Summary of the interview protocols for new patients, experienced patients and researchers

RESEARCHERS	EXPERIENCED PATIENT DELEGATES	NEW PATIENT DELEGATES*
• How important is patient participation in OMERACT for you? • Can you describe how you see the role of a patient participant at OMERACT? • For which research phases do you think patient participation is most appropriate? • What type of research do you think benefits most from patient participation? • What do you see as the greatest achievement of patient participation in OMERACT? • What barriers have you experienced related to incorporating the patient perspective in OMERACT initiatives? • Do you have suggestions how OMERACT could support researchers to optimize patient participation?	• What do you remember about your first OMERACT conference? • What makes OMERACT worthwhile to attend for a 2nd /3rd /4th time? • Which topic did you find most relevant to contribute to and which the least? • How do you see your role at this conference? • How could patients be of any help in studying "remission" and "flares"? • What do you see as the greatest achievement of patient participation in OMERACT? • What has been supportive to for you to participate in OMERACT? • What barriers have you experienced to contributing fully? • Do you have suggestions to optimize the input of patients in OMERACT?	• Can you briefly describe how your rheumatic condition has influenced your personal life? • How were you invited for this conference? • How do you see your role at this conference? • Do you have any idea how patients could be of help in studying "remission"? • Do you have any idea how patients could contribute to the research on "flares"? • Do you anticipate any barriers to your participation? • How could OMERACT support you to overcome barriers or to make you feel more confident? • Can you tell me, what are the most important things you hope to achieve by your participation at this OMERACT meeting?

* For new patient delegates we composed three versions: One protocol for the interview before the conference, one for half way, and one for after the conference.

time (SC) joined the research team. Codes were compared and discussed during several face-to-face meetings, resulting in sets of main categories. To reduce the risk of observer bias[21 22] all authors, representing different backgrounds, randomly cross-checked the coding and confirmed that there were no significant discordances. Triangulation took place by synthesizing data derived from the document analysis, interviews and participant observation.

Table 2 Main findings of the study

IMPACT	BARRIERS AND FACILITATORS
• Long term involvement of patients as collaborating partners has significantly influenced the OMERACT research agenda.	• The role of the leadership has been pivotal in implementing structural participation.
• The identification of fatigue as an under-researched domain and the increased research in this area would not have happened without the direct participation of patient research partners.	• Strict selection, training and individualized support of patient research partners are important facilitators for successful inclusion of the patient perspective in outcome research.
• Patient participation in OMERACT has stimulated the inclusion of the patient perspective in the development of core domain sets, outcome measures and other clinical concepts in rheumatology such as flares and remission.	• Like other new delegates patient research partners have to learn the objectives, procedures and culture of OMERACT before they can fully participate and contribute.
• Patient participation has been gradually embedded in the vision, culture and procedures of the OMERACT conferences.	• The interactive and inclusive conference design, together with a facilitative moderation style, are advantageous for the participation of patients.
• Patient participants provided a reality check for researchers.	• The intensity of the program and the academic terminology are important barriers for patients to participate fully.
• Patient participation in OMERACT has prompted new initiatives in other areas of health research to foster the inclusion of the patient perspective.	• Resistance to change and skepticism among researchers hinder full implementation of patient participation and increase chances of tokenism.
	• Doubts about the representativeness of the patient group still cause reluctance among professionals.

RESULTS

Influence of patient participation

Widening the research agenda

The main findings are shown in Table 2. There was broad consensus about the significant influence of patients on the research agenda by identifying previously neglected outcome domains such as sleep disturbances, flares and particularly fatigue.[23-26] The increased research in the domain of fatigue has been reported unanimously as the most illustrative result of this process which would not have occurred without the structural involvement of patients being listened to by receptive researchers. It has led to extensive studies into the nature, impact and measurement of fatigue in RA.[27-31] In 2006, fatigue was added to

the RA core-set as a recommended outcome for clinical trials.[32 33] More powerful instruments for measuring fatigue have been devised and validated[34 35] with the active involvement of patients.[36] The issue of fatigue was not new for rheumatologists.[37-39] During OMERACT 3 (1996) delegates carried out a ranking exercise where fatigue was already identified as a major concern for patients.[40] Eight measurement instruments were given for fatigue.[41] However, after this workshop, nothing happened for six years. Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only hearing from patients at OMERACT 6 made them change their perception of fatigue as an important outcome.

Core sets and outcome measures

Patients have identified domains that are relevant for disease-specific core sets for psoriatic arthritis, fibromyalgia, gout and vasculitis. Furthermore they contributed to the development of core sets for clinical concepts such as minimum clinically important difference (MCID) and remission, and played an important role in the assessment of the feasibility of core sets and outcome measures, one of the key components of the OMERACT Filter.[42] Patients have also been helpful in the development of PROs in the field of work productivity, adverse events, flares and psychosocial interventions.

Changing perspectives

Long term engagement with patients has changed the culture of OMERACT and the perceptions of its participants. Initially some researchers were concerned about inviting patients to the conference. They feared that patients were not able to transcend their personal experience, to generalize and to understand the rigor of the methodological discussions. These researchers became less reserved when, after a while, patients started to organize themselves and learned the OMERACT procedures, and they started to see the benefits. They confirmed that it changed their way of thinking and talking. Patients improved communication and brought dynamics to the dialogue because they were motivated, constructive and had no personal agenda. When discussions became extremely technical, patients reminded participants of the common goal of the conference by providing the human face of a person living with the condition day by day. Their presence made participants more explicit about the objectives of sessions and more explanatory about terms and concepts under discussion. Together with a reduced use of jargon this ‘forced’ simplification resulted in fewer misunderstandings for everyone.

Patients attending OMERACT for the first time reported significant learning curves and a variety of personal benefits. In fact all participants learned from the contact with

other parties. During this process participants gained trust, respect and understanding. One interviewee mentioned 'a reality check' as an important benefit: For professionals patient participation offered the opportunity to check the relevance and scope of their research: Are we doing the right things according to patients and are we using the right tools and methods?

Consequences outside OMERACT

Patient involvement in OMERACT has stimulated more patient oriented health research in many countries. Patients returning home after the conference have introduced the concept of participatory research in local projects or established networks of patient research partners.[43-46] With the input from several OMERACT participants, the European League Against Rheumatism (EULAR) developed recommendations for patient involvement in scientific projects[6]. Inspired by the experiences of OMERACT the organizing committee of the 6th International Shared Decision Making conference decided in 2011 to officially invite and facilitate for the first time four patient participants.[47] In the same year OMERACT delegates, patients as well as researchers, participated in the 2nd Core Outcome Measures in Effectiveness Trials (COMET) conference, demonstrating how the OMERACT methodology can be used in other disease areas.[48]

Facilitators

Leadership

The patient role evolved from a single focus group in 2002 to full participation in all parts of the conference in 2012. The long term commitment of the OMERACT organizing committee has been an important facilitator for this process. The organizers were intrinsically motivated to foster patient participation in the conference as they believed that patient input is essential to decide on the right measurement tools.[49] The decision to invite patients was not taken lightly, but once OMERACT leaders were determined, they wanted to do it properly. They provided full support to their designated patient participation leader and guaranteed funds for patients to attend the conference. The positive contributions made by patients were regularly acknowledged and stimulated OMERACT in 2006 to formulate basic principles for patient participation. This internal policy document represented a milestone by making patient participation part of the vision of OMERACT, embedded in a set of operational procedures. In 2011 OMERACT

decided on the principle that active involvement of patients would be a prerequisite for working groups suggesting themes in the OMERACT program.[12]

Patient selection

Since 2004 OMERACT has aimed for 10% of conference participants to be patients with a mixture of conditions, geographical spread and OMERACT experience. From the start it was clear that patients were expected not to represent any advocacy group or organization. Although they might be active in patient organizations, they should adopt a strict individual role at OMERACT. To avoid self selection recruitment is done through the clinic of participating physicians who proved to be ideally positioned to identify eligible patients that are able to make a valuable contribution[12]. A potential disadvantage of this is the possibility that patients feel unable to decline an invitation to please their consultant or to prevent jeopardizing their care.

Conference design

Constructive engagement with patients was enhanced by the small size of the conference, the open mind of the delegates and the spirit of OMERACT as a consensus-oriented initiative based on multi-stakeholder involvement. From the start patients have felt 'part of the family.'[50] The conference layout and the focus on active interaction and debate in small groups[10] have been facilitators for accepting and incorporating new perspectives. Some of the patients, all of whom were officially invited as full delegates with voting rights and access to all sessions, felt equal to professionals and acted as co-researchers. A majority of the patients saw their role as giving information or as an advisor. In particular new partners initially felt the same unequal relationship towards professionals as is experienced in clinical practice. The interviews revealed that new researchers and research fellows also experienced these disparities sometimes. However, participants who had attended several conferences confirmed that inequalities disappear over time.

Support

According to the organizers of OMERACT the first conference with patient involvement was actually the most easy and successful because there were no expectations. Partners formed a homogeneous group and the program was not demanding. Despite the fact that support was limited, patients felt welcome and accepted.[50] During subsequent conferences partners were extremely motivated to learn the OMERACT way and started to organize themselves. They formed a patient liaison group and developed introduction packs, information modules to bring newcomers up to speed, and a glossary in lay

language.[51] In 2010 new patients were matched with an experienced patient who took on the role of a buddy. To prevent overburdening, personalized programs were developed that helped patients to pace themselves and to prioritize sessions to attend. Finally, patients have urged that they be involved in the pre-conference work, feeling that early participation in working groups will best prepare them for worthwhile participation during the conference.

Facilitative moderation style.
The breakout discussion sessions form the heart of OMERACT and the quality of the moderation emerged as of utmost importance for patients to contribute to their full potential. From the perspective of patients an interactive and encouraging moderation style that fosters mutual learning through open dialogue was seen as an important facilitator to feeling confident to provide input. Sometimes patients were confronted with an apparently patronizing attitude of a moderator by whom they felt ignored or disqualified. Language was an underestimated barrier in discussion groups for those whose first language is not English. Some patients tended to hold back when they were afraid of not using the right words or expressions.

Barriers

Overburdening
OMERACT is an overwhelming experience for all patients attending the conference for the first time. Before the conference they felt privileged to be invited but insufficiently prepared and uncertain about what was expected from them. They had anticipated a learning experience and hoped to be able to make a difference. In fact most of them experienced the conference program as physically and mentally challenging. They had to adjust to the use of medical jargon and scientific terminology, and they were not always familiar with the rules for clinical research. The (intercontinental) travelling, the early and late starting times of sessions, and the duration of the conference increased the risk of overburdening. In particular new patients often participated beyond their physical capacities. Dealing with hierarchical power relations and strongly opinionated professionals was experienced as mentally challenging. A recurring barrier reported by patients was the lack of feedback on provided contributions. At times they felt that their experiential knowledge was not accepted as a valid source for scientific research, nor seen as relevant compared to the expert knowledge of professionals. Experienced patients coped better with the requirements of the conference. After the conference almost all

patients described their participation as having been a valuable learning experience. Although new patients presumed that they had not been very productive, they expected their contribution would be more effective at future conferences. Experienced patients as well as researchers confirmed that expectation.

Scepticism

In 2010 a few respondents believed that the disadvantages of patient participation outweighed the benefits. They judged the influence of partners out of balance and feared less interest in solving methodology challenges leading to a less diverse research agenda. They felt forced to engage with patients in areas of research where they saw little added value, such as imaging and biomarkers. Most of the patients confirmed this opinion and said that they thought they could offer little benefit to domains that were distant from their daily life. They believed that they could contribute most to research focused on their own condition and were uncertain about providing meaningful input during sessions dedicated to other conditions. To a great extent physicians agreed with this.

Lack of diversity

Institutionalization of patient participation gave rise to a debate regarding the representativeness of the patient group. OMERACT encouraged patients to provide the 'naïve' dimension of being familiar with the disease although there has been a tendency to raise the selection criteria in order to recruit higher educated patients. A small number of respondents objected to the participation of patients who do not understand the rigor of scientific research, slow down the process and hence cause irritation during breakouts. They argued that the patient perspective should be obtained during the pre-conference research work. Other participants emphasized the irreplaceable value of new patients bringing in authentic experiences, crucial for generating new ideas. They reported potential risks of patients becoming too professional, identifying themselves with researchers, and finally losing touch with their peers. Some respondents warned that patients who started acting as advocates might compromise the outcomes of OMERACT as a data-driven conference.

Discussion

Our study has proven that building patient participation structurally into the OMERACT conference program has guaranteed that themes that are important for patients were not only added to the research agenda, but have increased a sense of urgency and prompted sustainable collaboration in new research programs. From the example of fatigue we

learn that it takes more than a decade to identify a new domain of interest for patients, develop validated outcome measures and design intervention trials that target that new domain as a primary outcome. Patient involvement in OMERACT has enriched the research agenda and changed the perceptions of all stakeholders involved. Thus patient participation in a research conference is effective and complementary to other structured and deliberate procedures for agenda setting used in other disease areas.[52] In research agenda setting, various models and strategies have been developed with the help of different methods;[53] these may vary from more or less structured approaches, or from one-time consultation to regular advice from beginning to end. We see similarities between OMERACT and the Dialogue Model for agenda setting with patients, developed in The Netherlands: a stepwise approach for articulating patients' priorities in homogeneous groups followed by a dialogue with other stakeholders.[54-58] The Dialogue Model strives toward equal partnerships among patients and researchers in which control over the process is shared. In dialogue participants listen, try to understand each other and find a common ground.[59] Participants engage in a mutual learning process leading to a new perspective that is acceptable and recognizable for all involved.[60] The strength of OMERACT has been the vision that patient participation is an integral part of the conference which requires extensive support and continuous reflection by all stakeholders.

An important additional finding of this study is that patients and researchers feel patients' knowledge and competences could be even more fully utilized. Based on the results of this study and using the five categories of the FIRST model: Facilitate, Identify, Respect, Support and Train,[5] we have produced a list of recommendations that should be taken forward for consideration during future OMERACT conferences (Table 3). We summarize the remaining challenges for OMERACT as: (1) to study how the experiential knowledge can be better incorporated in the discussions during breakouts. It is known from the literature that patients' views are often excluded unintentionally[4 61] and OMERACT should develop more effective inclusion strategies; (2) to facilitate patient participation in working groups that meet between conferences to better prepare patients for their attendance at the conference; (3) to identify type and phases of research that are likely to benefit more from full involvement of patients; (4) to start reporting on the benefits and limitations of patient involvement; and (5) to guarantee a better representation of patients by inviting patients from continents that are currently absent.

Although we refer to examples outside OMERACT where conference organizers have taken notice of the way OMERACT has organized structural involvement of patients, the transferability of these recommendations may be limited to the specific context in which

Table 3 Recommendations for improving patient participation at future OMERACT conferences structured according to the FIRST-model

FACILITATE
• OMERACT should strive for structural involvement of patients in all phases of research, including pre- and post-conference activities. • Patients should receive personalized programs to prevent over-burdening. • Moderators should create conditions for participation by adapting the objectives and layout of breakout sessions. • Patient delegates should be involved in the site visit of the accommodation to assess accessibility.
IDENTIFY
• Selection criteria for patient research partners attending OMERACT conferences should be made explicit and should take into account the competence of the English language and experience in working with academic researchers. • OMERACT should strive for a more representative patient group regarding ages, continents and cultures. • Recruitment through the clinics of participating researchers should be continued. • OMERACT should differentiate between types of research regarding the desired level of involvement of patients.
RESPECT
• More attention is needed for information and acknowledgement after the conference. • Regular evaluation and reflection on the process of patient participation could improve the implementation and stimulate future research of its conditions and impact.
SUPPORT
• Patients should receive personalized guidance for reading the pre-conference materials. • Involvement in preparatory work is seen as the best way to prepare for the conference. • New patients appreciate to meet as a group once a day to exchange experiences. • Patient buddies should receive a clear briefing how they can support new patient participants.
TRAIN
• Patients need additional training on site to integrate and to achieve a minimum level of understanding OMERACT. • Training should focus on the expected role and tasks of patients, the topics on the conference program and only a minimum on bio-statistics.

the data have been collected. They have been formulated by the authors and have not gone through a thorough consensus process. This should be taken forward during future OMERACT conferences. Furthermore, based on the contingency of our findings we cannot provide a simple template for patient participation in all conferences.

Nevertheless, there may be lessons that other researchers can apply to the *process* of developing patient involvement.

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